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A novel, non-invasive approach in management of inflammatory dentigerous cyst in young child: A case report and review of literature

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ABSTRACT

Introduction: Developmental and inflammatory are the two variants of dentigerous cyst which is the second frequently reported odontogenic tumour. Displacement, mobility of tooth, swelling are the clinical characteristics. **Case report:** A six year old child reported to the department of Paediatric dentistry with history of swelling since 2-3 months in maxillary right anterior region, associated primary lateral incisor & canine were non-vital. Orthopantomogram showed well defined radiolucency in apical region of #53 region, deviated path of eruption of permanent canine. On fine needle aspiration straw coloured fluid was collected. Diagnosis of Inflammatory dentigerous cyst was done. **Treatment:** Primary maxillary right lateral incisor and canine were extracted. This was followed by marsupialization through the extracted socket by novel, non-invasive and cost-effective approach successfully. After nine months of follow-up; new bone deposition and change in path of eruption of canine was observed.

Keyword: Cyst, Marsupialization, Preservation, Tooth bud

1. INTRODUCTION

Cyst can be defined as pathological cavity which may or may not be or partially lined by epithelium and contains fluid, semisolid or gaseous material but not pus (Shear & Speight, 2007). Among the various odontogenic cysts associated with oral cavity, dentigerous cyst is the second most reported, approximately 20–24% (Murakami et al., 1995; Jena et al., 2004). Dentigerous cyst simply means sac containing or surrounding the tooth (Isser & Das, 2002). Developmental dentigerous cyst (DDC) and inflammatory dentigerous cyst (IDC) are the two types (Benn & Altini, 1996).

Accumulation of fluid or by generating a space between the reduced enamel epithelium and the enamel around the crown of unerupted tooth



causes expansion of follicle which originates in formation of cyst and thus results in impaction of tooth (Jena et al., 2004). Build up in inflammatory cells and desquamated epithelial cells into the cyst lumen increases osmolality of cystic fluid resulting in expansion of cyst. Also in some instances, the recurrent periapical inflammation from an overlying primary tooth extends into intra-follicular space cultivating into the dentigerous cyst (Clauser et al., 1994).

Mechanism of development of DDC is due to collection fluid between reduced enamel epithelium and unerupted tooth. Benn and Altini suggested three possible etiological mechanisms for occurrence of IDC are- 1) infection from periapical inflammation of non-vital primary tooth spreading and infecting dentigerous cyst, 2) radicular cyst associated with primary tooth spreading to involve follicle of unerupted successor, 3) inflammatory exudate of a nonvital deciduous tooth from apical and surrounding area infecting the follicle of an unerupted permanent tooth germ (Benn & Altini, 1996). The type of dentition involved in IDC is mixed dentition whereas DDC is permanent dentition. Mandibular third molars, maxillary canines, and mandibular premolars are the typical sites for occurrence of DDC (Clauser et al., 1994) but IDC is most commonly reported in mandibular premolar region as the roots of primary molars are in close association with the follicle of developing premolars which facilitate rapid spread of infection (Mintz et al., 2001).

Inflammatory type of dentigerous cyst is reported between ages of 6 to 12 years commonly whereas the developmental type of dentigerous cyst is common in 2nd to 3rd decade of life. IDC has high prevalence in mandible as compared to maxilla with ratio of 10:1 (Clauser et al., 1994; Kozelj & Sotosek, 1998) but high male and maxillary jaw predilection was observed in Indian population (Kalaskar & Kalaskar, 2016). Patient may not notice cyst unless and until swelling will cause pain or discomfort. These cysts are generally diagnosed on routine intra-oral periapical radiograph (IOPA) or Orthopantomogram (OPG). If the size of cyst measures more than 2 cm clinically; enlargement, misplaced teeth (displacement), mobility and sensitivity are observed (Bodner et al., 2003). IDC is generally associated with carious or non-vital primary teeth whereas DDC is associated with sound primary or permanent tooth (Bloch, 1928). Radiographically, carious tooth or teeth involving pulp or endodontically treated tooth along with a well-defined unilocular radiolucency with embedded crown of unerupted tooth along with a sclerotic border are the classical features of dentigerous cyst (Ziccardi et al., 1997).

IDC histologically shows presence of fibrous wall which is lined by non-keratinized stratified squamous epithelium of myxoid tissue, odontogenic remnants and rarely presence of sebaceous cells (Tuzum, 1997). Differential diagnosis involves radicular cyst, odontogenic keratocyst (OKC) - recently named as the keratocystic odontogenic tumour (KCOT) and Ameloblastoma. Radicular cyst is very rare in association with primary tooth. It is smaller in size and does not cause bony expansion and destruction but can be seen in case of dentigerous cyst (Shafer et al., 1994). Ameloblastoma is generally multilocular on radiograph; in case of unilocular it is exceptionally common in first decade of life (Shafer et al., 1994; Neville et al., 2002). KCOT is very aggressive lesion with high recurrence rate and is rare in first decade of life. In syndromic patients, KCOT may be detected in the first decade of life (Bresler et al., 2016).

Cystic contents in dentigerous cyst are usually straw coloured whereas in radicular cyst it is brown colour and in OKC it is dirty white cheesy material is reported (Sridevi et al., 2015; Nainani & Sidhu, 2014). Treatment includes enucleation, marsupialization, curettage, radical resection or combination of these techniques depending upon age of patient, size and extension of cyst, neighbouring vital structure, malignant transformation and mental health. In enucleation complete amputation of the cyst along with the lining occurs and healing is by primary closure, but in marsupialization the cyst is converted into a cavity which has opening to drain its cystic contents (Riachi & Tabarabi, 2010).

Marsupialization is mostly the chosen treatment for dentigerous cysts in mixed dentition as it results in preservation of developing permanent tooth buds thus preserving future occlusion (Bozdogan et al., 2011). If the cyst remains untreated, it may cause tissue destruction and deformities (Riachi & Tabarabi, 2010). This case presents a successful treatment option of non-invasive, novel Marsupialization in mixed dentition preserving underlying permanent tooth bud.

2. CASE REPORT

A 6yr. old boy reported to out patient department of Pediatric & Preventive Dentistry with chief complaint of swelling in upper right anterior region of jaw since 2-3 months and increasing in size. Intra-oral examination revealed presence of mixed dentition and soft, fluctuant swelling was present in labial sulcus extending from distal aspect of permanent maxillary central incisor (#11) to mesial aspect of Primary first molar (#54) (Fig.1a). Medical history was non-contributory. Patient had not visited any dentist / doctor or taken any medication for swelling. Along with swelling root pieces of maxillary primary right and left central incisors (#51,61), maxillary right lateral incisor (#52), maxillary right canine (#53), over-retained palatal root piece of primary first molar (#54), labial caries with # 63,74,83 whereas occlusal caries was present with #74,84. Orthopantomogram (OPG) was advised which showed 2 x 3 cm

well defined radiolucency in apical region of #53 region, mesially extending from distal aspect of developing #12 till mesial aspect of displaced tooth bud of #13 (Fig.-2). It was also observed that due to the radiolucency, tooth bud of permanent right maxillary canine (#13) was distally drifted almost touching the follicle of developing second premolar (#15) (Fig.-2). Based on clinical and radiographic findings provisional diagnosis dentigerous cyst was done.

Differential diagnosis involved radicular cyst, Keratocystic odontogenic tumour (KCOT, formerly OKC), and ameloblastoma. For confirmatory diagnosis Fine needle aspiration cytology (FNAC) was advised and performed after obtaining parents' consent. Area of interest was disinfected with local application of tincture iodine solution, 2% lignocaine gel was applied on the mucosal surface, 10ml 21 gauge disposable syringe was introduced in the lesion and plunger was pulled back for collecting the contents of the cavity. The needle was moved through the lesion three or four times in different directions, straw coloured fluid was collected which suggested dentigerous cyst (Fig.1b). Immediately after FNAC, size of the lesion reduced. FNAC report showed cholesterol crystals having a broken cover glass appearance and presence of inflammatory cells mainly lymphocytes and RBC's suggestive of IDC. Parents were informed about both treatment options along with their advantages and disadvantages. But keeping in view the age of child and for preservation of tooth buds of permanent maxillary teeth, parents were advised marsupialization. Parents also readily gave consent for marsupialisation. As marsupialization was the planned further confirmatory histological analysis could not be performed.

Local anaesthesia was obtained by using 2% lignocaine with adrenaline (1:80,000, 2% Lignox®) buccally with infra-orbital block, and anterior palatine block, extraction of teeth # 51,52,53,61 was carried out and cystic content drained from the socket. Curettage taking care of underlying permanent tooth buds and porch was created through the extraction socket of #53 (Fig.1c). Copious irrigation of cystic cavity with diluted solution of tincture iodine (Betadine®) with saline was carried out. Dressing of tincture iodine damped gauze was placed in the cavity (Fig.1d). As patient was young and care of the wound may not be possible increasing the chances of infection, we decided to change the dressing every 48 hrs till complete healing occurs. Follow-up OPG was also advised initially every month and later every three months. At end of nine month follow-up, eruption of #12 was observed clinically (Fig.1e) and OPG showed complete healing of radiolucency and change in path of eruption of #13 (Fig.3). No recurrence was observed during this follow-up period and follow-ups are ongoing till the complete eruption of permanent dentition.



Fig 1a. Intraoral swelling present in the area of # 52 till # 14



Fig 1b. FNAC showed presence of strawberry colored liquid



Fig 1c. Porch created through the extracted socket of # 53



Fig 1d. Dressing given through the extracted socket



Fig 1e. After 9 months normal eruption of # 12 is observed

Figure 1 Intra-oral photographs pre-operative, operative & follow-up picture

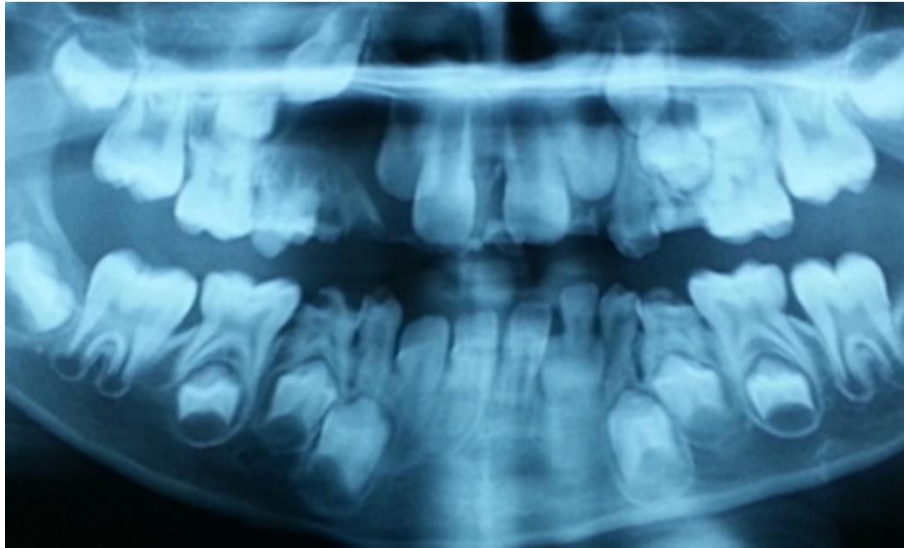


Figure 2 Pre-operative Orthopantomogram (OPG) showing radiolucency in maxillary right anterior region with carious # 52, 53 and deflected path of eruption of # 13.



Figure 3 OPG after 9 months of follow-up of marsupialization showing complete healing of radiolucency in maxillary anterior region and normal path of eruption of #13.

3. DISCUSSION

Oozing of cystic contents either during an extraction of a primary tooth or decompression is a confirmatory clinical feature of the cyst (Kozelj & Sotosek, 1999). KCOT, Dentigerous cyst and radicular cyst are the most common cyst found in children and adolescent (Nannan et al., 2014). Developmental and inflammatory are the two types of dentigerous cyst. Piling up of fluid either between the reduced enamel epithelium and the enamel or in between the layers of the enamel organ sources the developmental type of dentigerous cyst. Whereas Bloch proposed that persistent periapical inflammatory exudates from the overlying primary non-vital tooth reaching out to the follicle of the unerupted permanent successor causes the initiation of inflammatory type of dentigerous cyst (Bozdogan et al., 2011; Main, 1970; Toller, 1970).

IDC has high prevalence for mandibular arch (Clauser et al., 1994) but Indian population IDC has shown high incidence in maxillary arch (Kalaskar & Kalaskar, 2016) which was coincided in present case also. IDC is detected in mixed dentition commonly, age ranges from 6-12 years (Kozelj & Sotosek, 1999), present case was also reported in early mixed dentition and age of child was six years which coincides with previous literature. Non-vital carious tooth, endodontically treated tooth or trauma were etiological finding of IDC whereas developmental dentigerous cyst is associated with non-carious tooth (Bloch, 1928), here in above case

carious nonvital #52, 53 were present favouring the diagnosis of IDC. Diagnosis depends upon Clinical & Radiographic features but FNAC and histopathologic examination of the cyst contents and lining are most important for final diagnosis.

Differential diagnosis of IDC involves radicular cyst, KOCT/OKC, Ameloblastoma. In our case, child was in his first decade of life, non-syndromic, associated tooth was carious & non-vital, lesion was slow growing, straw coloured fluid was collected on aspiration, content of fluid had 6-7 gm/100ml protein, drooling of fluid through the extraction socket favoured the diagnosis of inflammatory dentigerous cyst. The size & location of the cyst, amount of bone present and adjacent vital structures are the factors which influence the choice of treatment of any cyst. If the radiographic size of cyst is less than 5 cm and cyst may be unilocular or multilocular, enucleation or marsupialization are the treatment choices advised. If this option does not result in healing or recurrence occurs radical resection is advised (Nyimi, 2019). If the radiographic size of cyst is more than 5cm & cyst is unilocular or multilocular and presence of multiple cortical perforations, involvement of adjacent tissue, recurrence and malignant transformation is observed radical resection is advised (Nyimi, 2019).

In the Partsch I procedure, a window at least 1 cm in diameter is made in the cyst lining to the oral cavity and Partsch II is enucleation (Dowsett, 1931). In Marsupialization or decompression or the Partsch I operation, intra-cystic pressure is reduced by establishing a surgical pathway in between cyst and the oral cavity for draining out the cystic contents. In maxillary arch, this window would have opening be either in into the oral cavity or maxillary sinus or nasal cavity but in mandibular arch it has intra-oral window (Kozelj & Sotosek, 1999; Bloch, 1928; Ji et al., 2012). In our case, the size of cyst was about 6 cm, no involvement of adjacent tissue or nerve, no malignant transformation was present and no history of previous history or treatment promoted conservative treatment choices- enucleation or marsupialization. Enucleation would have resulted in sacrificing permanent lateral incisor and canine, and prolonged use of removable denture would have resulted in bone resorption which would further compromised the final implant supported prosthesis as child was in his first decade of life, marsupialization was the treatment of choice as it promoted the preservation of future dentition and positive psychological impact on child.

Many methods or devices are recommended or used to keep patency, these include gauze soaked in iodoform and bacitracin ointment, customized acrylic obturator, a space-maintaining appliance with a resin projection, and a modified removable partial denture a nasopharyngeal anaesthesia tube, modified pediatric airway device, modified tubes as decompression catheters, polyethylene tube of various length, modified catheters made from an intravenous (IV) administration set (injection site or spin-lock connector), nasal cannula (O₂ connector), and the funnel end of an all-purpose urethral catheter etc. (Jiyeon et al., 2017). Use of obturator, modified RPD was not used as present case was in developing age and these could be interfered with eruption of permanent lateral incisor gauze soaked with Povidone-iodine (PVP-I) (Betadine®) solution which was changed every 48 hrs. PVP-I is efficient for neutralizing both gram-positive and gram-negative microorganisms as well as against *Candida albicans* (Bogash, 1956; Gershenfeld, 1957), it is non-toxic to surrounding tissue & easily accepted by children so we decided to use it.

The drawback or limitation of marsupialization that cyst lining is not obtainable for histological diagnosis, patient's co-operation is needed to keep maintain the oral hygiene specially operated area till purpose of the treatment is achieved, prolong treatment duration as compare to enucleation or resection, difficult in special child / person as co-operation is not present, may not be successful in large cyst. In these instances, enucleation or radical resection may be the alternate choice. In our case, we could achieve the goal of preserving the permanent tooth buds and healing of the cystic cavity with non-invasive technique of marsupialization with positive impact on child's psychology.

4. CONCLUSION

Inflammatory type of dentigerous cyst is frequently reported cyst in children, leading to swelling, displacement of teeth and malocclusion even impacting the jaw growth and development in some instances. Thus, affecting the functions and aesthetic of the patients; leading to low self-esteem. Treatment planning is governed by the factors like the size and location of the cyst, vicinity of important structures like nerve and tooth germs, and age of the patient. With early & proper diagnosis, treatment planning and patient's co-operation, not only healing of cyst but also preservation of developing permanent tooth bud can be executed thus maintaining future occlusion with a positive impact on the child's esteem.

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Conflict of interest

The Authors have no conflicts of interest that are directly relevant to the content of this clinic-pathological case

Financial Resources

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Informed Consent

Oral and Informed Consent was obtained from the patient's parents.

Author's Contribution

All the authors contributed equally to this case report.

Data and materials availability

All data associated with this study are present in the paper.

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